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Gene Section

Review

WDR48 (WD repeat domain 48)

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Abstract

WDR48 is a WD40-repeat containing protein that regulates the activity of three human deubiquitinating enzymes: USP1, USP12 and USP46. The best characterized role of WDR48 is regulation of the DNA damage response in complex with USP1. The USP1/WDR48 complex promotes deubiquitination of monoubiquitinated FANCD2 and monoubiquitinated PCNA, which are required in the regulation of translesion synthesis (TLS) and Fanconi anemia (FA) DNA repair pathways, respectively. In addition, in complex with USP12 and USP46, WDR48 regulates other important cellular processes such as synaptic transmission or signalling through the Akt, Notch and T-cell receptor pathways.

Keywords
WDR48; USP1; USP12; USP46; DNA damage Fanconi anemia

Identity

Other names
P80, UAF1, SPG60, KIAA1449

HGNC (Hugo)
WDR48

Location
3p22.2 (Starts at 39051986 and ends at 39096671 bp from pter (according to hg-38))

Local order
Based on MapViewer, genes flanking WDR48 are:
-SCN10A (sodium voltage-gated channel alpha subunit 10); 3p22.2
-SCN11A (sodium voltage-gated channel alpha subunit 11); 3p22.2
-WDR48 (WD repeat domain 48); 3p22.2
-GORASP1 (golgi reassembly stacking protein 1); 3p22.2
-TTC21A (tetratricopeptide repeat domain 21A); 3p22.2

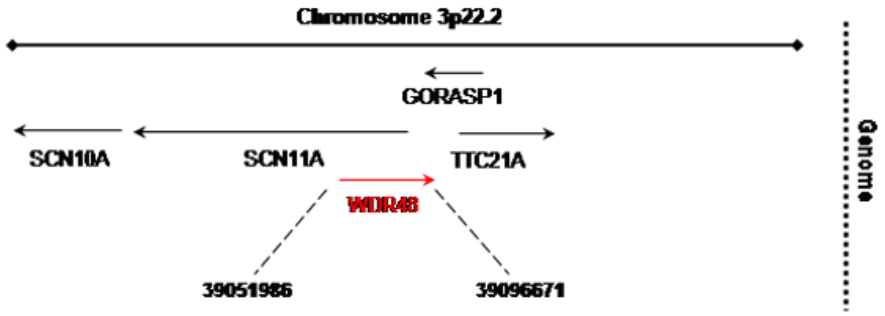

Figure 1: Localization of WDR48 on chromosome 3p22.2. WDR48 starts from 39051986 and ends at 39096671 bp.

DNA/RNA

Description

WDR48 gene is located on chromosome 3 in the region p22.2. WDR48 was first cloned in 2000 (Nagase et al., 2000) as a gene coding for a 607 amino acid protein called KIAA1449. In 2002, Park and co-workers (Park et al., 2002) cloned the full-length cDNA encoding the 677 amino acid WDR48 protein, which they called p80.

Transcription

Ten alternatively spliced transcript variants have been identified (WDR48-001, -003, -004, -005, -006, -007, -008, -009, -010 and -011) for this gene. Transcription variants -001 and -008 encode for proteins containing 677 and 162 amino acids, respectively. The remaining variants are not translated into a protein product.

Pseudogene

Not reported pseudogenes. Paralogs for WDR48 are STRN4 and STRN3.

Protein

Description

The WDR48 gene encodes a 677 amino acid protein (from the -001 splice variant) with a predicted molecular weight of 76,21 kDa. WDR48 belongs to the WD repeat (WDR) domain-containing protein family. This family is characterized by the presence of several copies of an approximately 40 residues-long sequence motif containing Trp-Asp repeats. WDR48 contains eight WD repeats (Figure 2). (Repeats WD1-WD7 adopt a 7-bladed β-propeller structure shaped as a toroid with a narrow central pore (Yin et al., 2015), whereas the WD8 repeat is included into the so-called ancillary domain. On the other hand, the C-terminus of WDR48 contains two SUMO-like domains (SLD1 and SLD2) that adopt a coiled-coil structure.

As further detailed below, WDR48 functions as a cofactor that stimulates the enzymatic activity of deubiquitinating enzymes (DUBs). In this context, the β-propeller domain of WDR48 mediates the binding to the enzymes, whereas the SLD domain contributes to the recruitment of different substrates (Cohn et al., 2007; Yin et al., 2015; Yang et al., 2011).

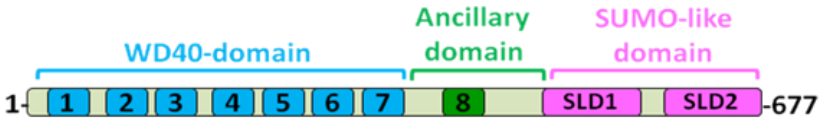

Figure 2: Structural organization of the WDR48 protein. WDR48 contains eight WD40 repeats. WD1-WD7 adopt a β-propeller structure. WD8 is part of the ancillary domain. The C-terminus contains two tandem SUMO-like domains (SLD1 and SLD2) that adopt a coiled-coil structure.

Expression

WDR48 is expressed in a wide variety of tissue types including testis, brain, thyroid, prostate, lung and bone marrow.

Localisation

Initially reported as an endosomal protein (Park et al., 2002), WDR48 is also detected in the cytoplasm. The nucleocytoplasmic distribution of WDR48 can be further regulated by interaction with other proteins, such as the human papillomavirus (HPV) E1 helicase or the human deubiquitinating enzyme USP1. These proteins bear active nuclear localization signals (NLSs), and facilitate WDR48 import into the nucleus via a piggyback mechanism (Cotée-Martin et al., 2008; Garcia-Santisteban et al., 2012).

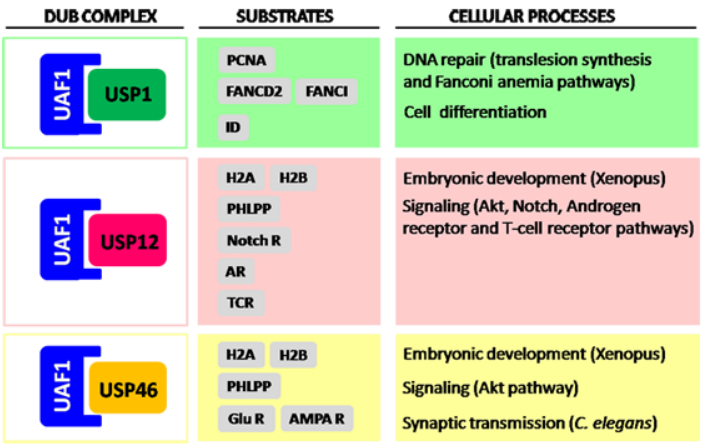

Figure 3: By activating USP1, USP12 and USP46 deubiquitinases, WDR48 regulates the ubiquitination level of several substrate proteins and takes part in a variety of cellular processes.

Function

The best-characterized role of WDR48 is as a cofactor of USP1, USP12 and USP46, three deubiquitinating enzymes belonging to the Ubiquitin Specific Protease (USP) family (Cohn et al., 2009). The intrinsic enzymatic activity of these DUBs is relatively low, and is increased by up to 30 fold upon complex formation with WDR48 (Cohn et al., 2007, Faesen et al., 2011, Yin et al., 2015). Through its β-propeller domain, WDR48 binds to the so-called "Fingers" domain of the DUBs (Garcèa-Santisteban et al., 2012; Olazabal-Herrero et al., 2015; Yin et al., 2015). The Fingers domain is far away from the catalytic site of these enzymes, and WDR48-mediated stimulation of DUB activity involves an allosteric mechanism. WDR48 binding induces a structural rearrangement of the DUB active site that increases catalytic turnover. (Villamil et al., 2012; Yin et al., 2015; Li et al., 2016). Of note, binding to WDR48 does not modify the affinity of the DUBs for their substrates.

By activating these three different deubiquitinating enzymes, WDR48 contributes to the regulation of the ubiquitination level of several substrate proteins, and thus plays a role in a variety of cellular processes (summarized in figure 3).

In complex with USP1, WDR48 regulates DNA repair by reverting the monoubiquitination of PCNA in the translesion synthesis pathway (Huang et al., 2006), and of FANCD2 / FANCI in the Fanconi anaemia pathway (Nijman et al., 2005; Cohn et al., 2007, Yang et al., 2011; Van Tweest et al., 2017). Moreover, the USP1/WDR48 complex promotes the repair of DNA damage by homologous recombination (Murai et al., 2011; Park et al., 2013). This has been reported to be, at least in part, mediated by the interaction of WDR48 withRAD51AP1 (Liang et al., 2016; Cukras et al., 2016) in a manner that could be independent from USP1 (Liang et al., 2016) (Figure 4). Besides these DNA repair-related functions, the USP1/WDR48 complex deubiquitinates inhibitors of DNA binding (ID) proteins and thus regulates differentiation in osteosarcoma cells (Williams et al., 2011).

In complex with either USP12 or USP46, WDR48 regulates embryonic development in a Xenopus model by deubiquitinating histones H2A and H2B, (Joo et al. 2011), and negatively regulates the Akt signaling pathway by stabilizing the PHLPP1 phosphatase (Gangula et al., 2013; Li et al., 2013).). Besides the Akt pathway, at least three other signalling pathways are regulated by WDR48 in complex with USP12. The USP12/WDR48 complex negatively regulates Notch signaling by stabilizing the nonactivated form of the Notch receptor (NotchR) (Moretti et al., 2012). Conversely, it positively regulates T-cell receptor (TCR) signaling by increasing the stability of the TCR signaling complex (Jahan et al., 2016), and it promotes androgen receptor (AR) signaling both directly (by deubiquitinating and stabilizing the AR protein) and indirectly (by reducing Akt-mediated AR inactivation) (Burska et al., 2013; McClurgh et al., 2104).).

Finally, in complex with USP46, WDR48 may contribute to modulate synaptic transmission in the brain by deubiquitinating and regulating the turnover of neuronal glutamate receptors (GluR) and AMPA receptors (AMPAR) (Dalhberg et al., 2014; Huo et al., 2015).

In addition to the cellular functions described above, WDR48 also plays a role in viral pathogenesis. In fact, WDR48 was originally identified as p80, a cellular protein targeted by the herpesvirus saimiri Tip protein during viral infection (Park et al., 2002). WDR48 has been subsequently found to bind other viral proteins, such as the E1 helicase of HPV (Cté-Martin et al., 2008) or the EBNA3C protein of Epstein-Barr virus (EBV) (Ohashi et al., 2015). These interactions, which may also involve the DUBs, appear to promote the replication and maintenance of the viral genome in HPV-infected keratinocytes (Cté-Martin et al., 2008, Lehoux et al.,

2014, Gagnon et al., 2015), as well as the growth of EBV-infected lymphoblastoid cells (Ohashi et al., 2015).

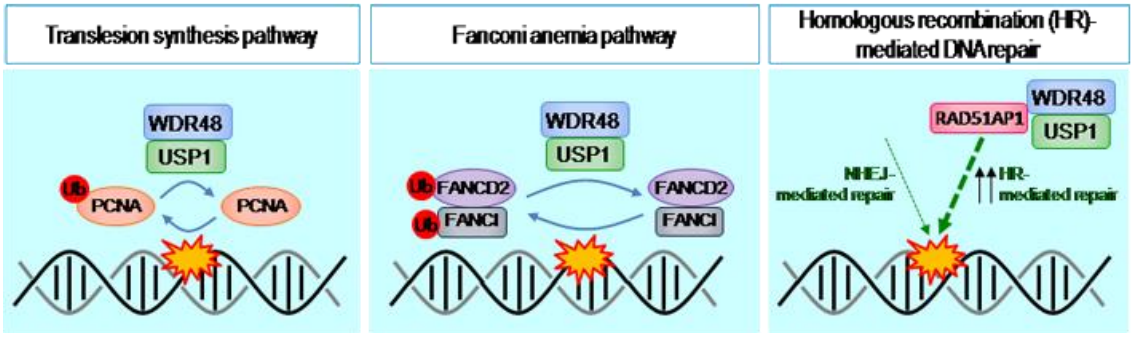

Figure 4: Summary of the roles of the USP1/WDR48 complex in the DNA damage response. USP1/WDR48 regulates DNA repair by reverting the monoubiquitination of PCNA in the translesion synthesis pathway and of FANCD2/FANCI in the Fanconi anaemia pathway. In addition, the USP1/WDR48 complex promotes the repair of DNA damage by homologous recombination by the interaction of WDR48 with RAD51AP1.

Homology

The WDR48 gene is conserved in chimpanzee, Rhesus monkey, dog, cow, mouse, rat, chicken, zebrafish, fruit fly, mosquito, C.elegans, A.thaliana, rice and frog.

Mutations

Somatic

A survey in the COSMIC mutation database (accession date: 17 November 2016) revealed a total of 71 mutations that lead to different modification in different human tumors. Most of the modifications are missense mutations whose functional consequences need to be addressed (Table 1).

Table 1: Cancer-associated mutations in WDR48. The table shows a detailed list of WDR48 cancer-associated mutations reported to date (November 2016) in the COSMIC mutation database, including the DNA modification (CDS Mutation), protein modification (AA mutation) and affected tissue. Missense amino acid substitutions are indicated in black, nonsense amino acid substitution in red and frameshift insertion/deletions in blue. Synonymous amino acid substitutions have been omitted.

CDS Mutation	AA mutation	Tissue
c.8C>T	p.A3V	Skin
c.35 36GG>>AA	p.R12Q	Skin
c.43G>T	p.V15L	Urinary tract
c.53C>T	p.S18F	Skin
c.56A>G	p.Y19C	Haematopoietic and lymphoid
c.64C>T	p.R22*	Endometrium
c.83A>G	p.Y28C	Large intestine
c.88C>T	p.R30*	Large intestine
c.113C>T	p.L45F	NS
c.148C>T	p.R50*	Large intestine
c.175G>A	p.V59I	NS
c.189G>T	p.K63N	Upper aerodigestive tract
c.206C>T	p.A69V	Stomach
c.221A>G	p.H74R	Stomach
c. 277G>T	p.A93S	Autonomic ganglia
c.284C>T	p.S95F	Prostate
c.290C>T	p.T97M	Oesophagus
c. 304T>C	p.W102R	Breast
c.317A>G	p.K106R	Oesophagus
c.330G>T	p.M110I	Skin
c.374_375insT	p.A126fs*4	Oesophagus
c.403G>T	p.A135S	Liver

c.504A>C	p.K168N	Large intestine
c.565G>A	p.E189K	Breast
c.585G>T	p.W195C	Stomach
c.610A>G	p.M204V	Haematopoietic and lymphoid
c.659G>C	p.R220T	Oesophagus
c.668C>T	p.T223M	Large intestine
c.682G>T	p.G228C	Small intestine
c.703C>T	p.R235C	Large intestine
c.745C>T	p.R249*	Large intestine
c.746G>T	p.R249L	Liver
c.770C>T	p.A257V	Prostate
c.784G>A	p.D262N	Cervix
c.859C>T	p.R287W	Skin
c.871T>G	p.C291G	Endometrium
c.916G>T	p.A306S	Endometrium
c.934A>G	p.I312V	Breast
c.1001C>T	p.A334V	Stomach
c.1006G>T	p.G336*	Liver
c.1007G>C	p.G33A	Urinary tract
c.1024T>A	p.C342S	Central nervous system
c.1036A>G	p.I346V	Upper aerodigestive tract
c.1102A>G	p.I368V	Haematopoietic and lymphoid
c.1210G>A	p.E404K	Cervix
c.1326G>T	p.W442C	Large intestine
c.1343C>A	p.A448D	Endometrium
c.1343C>T	p.A448V	Prostate
c.1345G>A	p.G449S	Pancreas
c.1434_1435insTG	p.V479fs*2	Biliary tract
c.1141C>T	p.P481S	Skin
c.1451A>G	p.E484G	Pancreas
c.1486C>A	p.Q496K	Haematopoietic and lymphoid
c.1495C>T	p.R499*	Endometrium
c.1508_1524del17	p.N504fs*10	Ovary
c.1522C>T	p.Q508*	Skin
c.1543G>T	p.V515L	Urinary tract
c.1567C>T	p.R523C	Skin
c.1682A>G	p.K561R	Large intestine and Pancreas
c.1705C>T	p.L569F	Skin
c.1743A>C	p.K581N	Lung
c.1740delA	p.D583fs*75	Biliary tract
c.1812T>G	p.I604M	Large intestine
c.1858G>A	p.E620K	Urinary tract
c.1870G>A	p.E624K	Large intestine and Urinary tract
c.1895C>T	p.A632V	Endometrium
c.1963C>T	p.R655*	Stomach
c.1969G>A	p.V657M	Skin
c.1989G>C	p.K663N	Lung
c.1999G>A	p.D667N	Skin
c.2017C>T	p.R673C	Breast

Implicated in

Fanconi anaemia

In mice, knockout of WDR48 results is a phenotype that resembles the phenotype of human patients with Fanconi anemia (FA) (Park et al., 2013), a rare genetic disease characterized by congenital malformations, progressive bone marrow failure, genomic instability, hypersensitivity to DNA cross-linking agents and increased susceptibility to cancer (Kee and DAndrea 2012).

Autosomal recessive spastic paraplegia type 60 (SPG60)

Hereditary spastic paraplegias (HSP) are rare neurodegenerative disorders. The main pathological

characteristic is the degeneration of the corticospinal tracts. Novarino and co-workers (Novarino et al., 2014) identified homozygosity for a WDR48 deletion mutant (c.1879_1881delAAG), resulting in a deletion of glutamic acid at position 628 (E628del), in a single individual from a consanguineous family that leads to spastic paraplegia type 60. The contribution of this variant to SPG60 has not been confirmed.

To be noted

Note

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